Recurred Ameloblastoma of the Maxillary Sinus, Coexisting in the Frontal Sinus : A Case Report

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－ ABSTRACT －

Ameloblastomas are rare tumors of the paranasal sinus, which are considered benign yet locally aggressive neoplasms in the vast majority of cases. Although ameloblastomas involving the maxillary sinus are often reported, cases involving the frontal sinus are rare. We present the case of a 45-year-old male patient with recurred ameloblastoma of the left maxillary and frontal sinuses. **(J Clinical Otolaryngol 2015;26:92-96)**

KEY WORDS : Ameloblastoma · Maxillary sinus · Frontal sinus.

Introduction

Ameloblastoma is a benign tumor of epithelial origin with invasive and destructive growth characteristics ; [1] distant metastasis is very rare. However, the recurrence rate is high because of incomplete surgical resection. The tumor grows slowly and the behavior of this entity is that of a low-grade malignant tumor.[2] Its radiological appearance generally varies, and the margins of the tumor are ambiguous. As a result, the tumor frequently recurs after surgical removal due to the difficulty of achieving complete surgical resection, especially in the paranasal sinus.[3-5] Ameloblastoma occurs most often in the mandible, and less commonly in the maxilla. Frontal sinus ameloblastoma is very rare. Herein, we report a rare case of maxillary ameloblastoma with frontal sinus recurrence, which requires wide excision.

논문접수일 : 2015년 3월 9일
논문수정일 : 2015년 4월 20일
심사완료일 : 2015년 5월 27일
교신저자 : 이영찬, 134-727 서울 강동구 동남로 892
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Case Report

A 45-year-old Korean man visited our hospital for treatment of nasal obstruction. He had undergone medial maxillectomy and radiotherapy due to ameloblastoma in Indonesia 4 years prior. The tumor did not recur for the first two years of follow-up based on magnetic resonance imaging (MRI) records obtained from an Indonesian hospital. On presentation to our hospital, he reported nasal obstruction, rhinorrhea, and hyposmia. Physical examination findings included a lateral rhinotomed scar resulting from prior medial maxillectomy ; cheek swelling and tenderness were not observed. Endoscopic findings revealed a mass occupying the left, anterior and inferolateral maxillary sinus. Computerized tomography showed a heterogenically enhanced mass in the left maxillary sinus extending to the alveolar process with anterior and lateral maxillary sinus wall destruction. MRI also revealed a heterogenous high signal area in the left maxillary and frontal sinus, which included the frontal recess (Fig. 1). Preoperative pathological examination done at another hospital revealed high-grade dys-

plasia; thus, a biopsy was performed via endoscopic surgery and intraoperative pathologic results were consistent with ameloblastoma of the maxillary sinus, sparing the posterior wall, frontal sinus, frontal recess, and inferior turbinate.

Under general anesthesia, the inferior partial maxillectomy was performed, with a lower part of the infraorbital foramen serving as an inferior margin. The nasal septum was saved medially by longitudinal cutting. After extraction of the lateral incisor, the maxillectomy was continued to the hard palate. We also performed endoscopic sinus surgery for excision of the frontal sinus mass (Fig. 2). The posterior margin covered the posterior wall of the maxillary sinus. Pathological examination of the excised lesion indicated left maxilla bony invasion and the frontal sinus, including the frontal recess, showed ameloblastoma atypia ; however, the posterior wall of the maxillary sinus exhibited only chronic inflammation. After the surgery, we confirmed recurred ameloblastoma through pathological analysis (Fig. 3). In addition, an obturator was prescribed to the patient as a prosthesis. The patient has been followed in an outpatient clinic for about a year without signs of recurrence (Fig. 4).

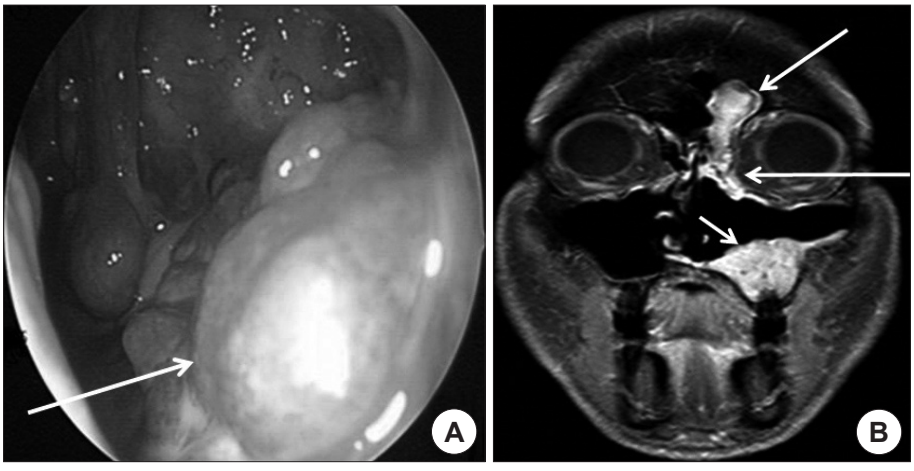

Fig. 1. Endoscopic & MRI findings. A : Endoscopic findings of the left maxillary sinus, anterior and inferior and lateral maxillary sinus occupying mass (arrow). B : Facial MRI findings of heterogenous high signal appearance in the left maxillary and frontal sinus, which included the frontal recess (arrow).

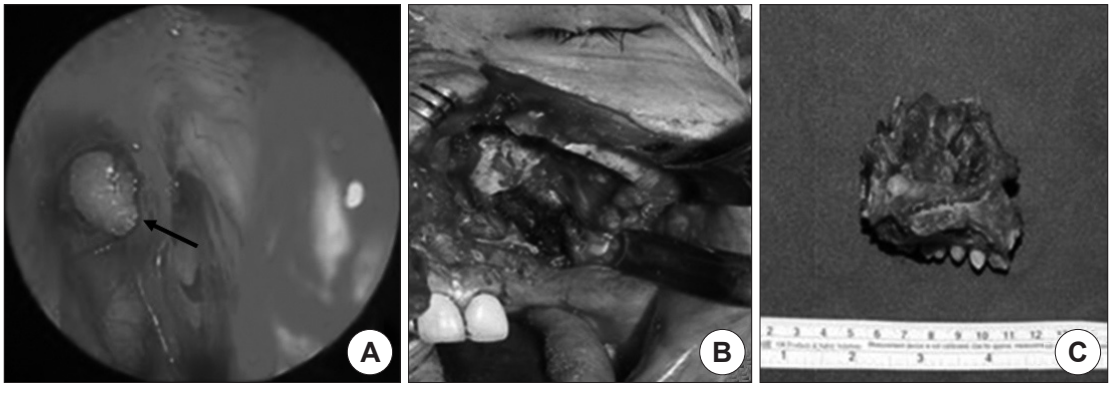

Fig. 2. Specimen during operation. A : Endoscopic findings of the left frontal recess mass during excision of ameloblastoma in the frontal sinus through endoscopic sinus surgery (arrow). B : This picture shows the mass excised through left maxillectomy. C : The excised gross specimen with left maxilla bony destruction.

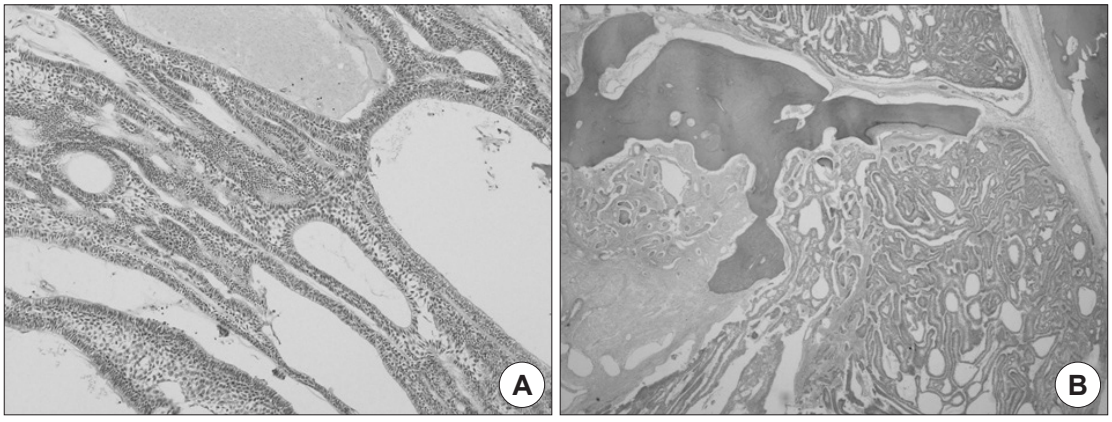

Fig. 3. A : Ameloblastoma infiltration of trabecular bone (H&E, ×20). B : The tumor shows peripheral palisading and central stellate reticulum areas (H&E, ×100).

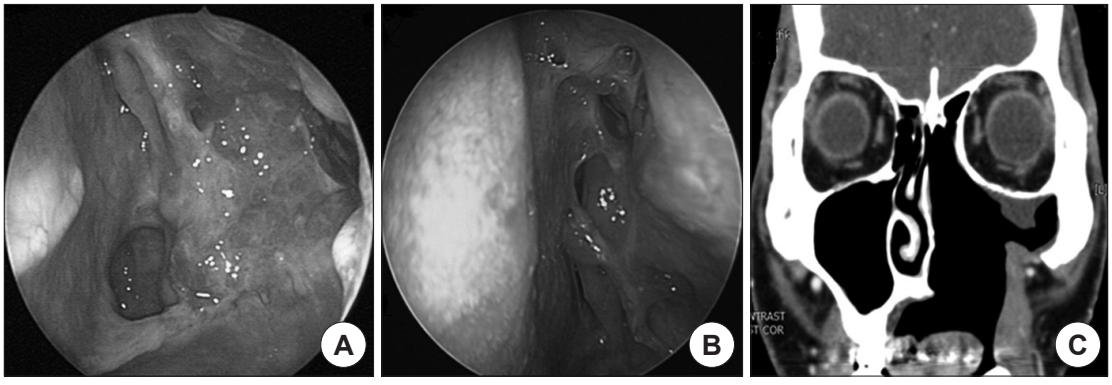

Fig. 4. Post operation 9 month findings. A : Follow up endoscopic findings of left maxillary sinus. B : Follow up endoscopic findings of left ethmoidal sinus & frontal recess. C : Follow up PNS CT enhanced coronal imaging.

Discussion

Ameloblastoma is a rare benign tumor of the maxillofacial and peripheral regions that most often occurs in the mandible. Maxillofacial ameloblastomas are currently believed to account for 20% of all ameloblastomas.[6] Most of these are maxillary sinus ameloblastomas. A similar case was reported for sinonasal ameloblastoma in 2011.[7] In addition, the abundant blood supply of the maxilla enables spread.[8] This case was important because it recurred in the maxillary and frontal sinus, including the frontal recess, but with a skipped region. The most common symptom of ameloblastomas in the maxillary sinus is painless swelling of the involved part of the jaw ; however, our patient did not have these typical symptoms. Pain is an infrequent finding, but it is not clear whether the pain is due to the tumor itself or a secondary infection. Nasal obstruction, localized facial swelling of the cheek, gingiva, and hard palate was commonly described in maxillary sinus ameloblastomas.[4] Our patient's chief complaint was nasal obstruction, and thus the intranasal mass was discovered through nasal endoscopy. In case of enhanced mass involved in the maxillary sinus, usually differential diagnosis from other malignant and invasive tumors such as craniopharyngiomas is needed.[9] However, our patient had a history of ameloblastoma in the same region.

A number of methods have been introduced in the

treatment of ameloblastoma, including wide excision, curettage, enucleation, cryotherapy, cautery, laser usage, radiotherapy, and chemotherapy. Excellent results have been obtained with radical excision and other methods.[6,8] It appears that the best surgical method for limited maxillary ameloblastoma is wide excision of the tumor with a 10- to 15-mm margin of normal bone, if possible ; [4] Therefore, we believe that the inferior partial maxillectomy was the best treatment choice in our case. However, the radical excision of the frontal sinus tumor was limited due to different endoscopic approach used compared to the removal of the maxillary sinus tumor. Due to these limitations, the results of endoscopic resection should be followed closely.

Finally, several pathologic types of ameloblastoma present in the maxilla. The unicystic type has a low recurrence rate as compared with the multicystic type, but this type has not been found in the maxilla.[10] The desmoplastic type has been identified predominantly in the anterior and posterior regions of the maxilla, despite very low incidence. These ameloblastomas have a low recurrence rate[6] although prognosis and behavior vary according to the various types in maxillary ameloblastomas. The maxillary ameloblastoma presented in this case was an atypical cell-type lesion, which recurred in the form of a frontal sinus ameloblastoma that included the frontal recess 4 years after the initial operation. Recurrence and extension of maxillary ameloblastomas are very rare.

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